RESEARCH REPORTS

Clinical

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Effects of Lip Revision Surgery in Cleft Lip/Palate Patients

ABSTRACT

The decision for lip revision surgery in patients with repaired cleft lip/palate is based on surgeons' subjective evaluation of lip disability. An objective evaluation would be highly beneficial for the assessment of surgical outcomes. In this study, the effects of lip revision on circumoral movements were objectively quantified. The hypothesis was that lip revision increases scarring and impairment. The study was a non-randomized clinical trial that included patients with cleft lip who had revision, patients with cleft lip who did not, and non-cleft control individuals. Three-dimensional facial movements were measured. Revision patients were measured before and after surgery. Other individuals were measured at similar intervals. Regression models were fit to summary measurements, and changes were modeled. Patients with repaired cleft lip/palate had fewer mean movements than control individuals. Lip revision did not worsen mean movements; however, individual patients' movements varied from 'improvement' to 'no change' to 'worse' relative to those of control individuals.

KEY WORDS: facial soft tissues, lip revision, movement, surgery, outcomes.

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INTRODUCTION

Many patients with a repaired cleft lip/palate require lip revision surgery for optimum esthetics (Bardach *et al*., 1984; Marsh, 1990; Cohen *et al.,* 1995). Children can have multiple surgeries of this type that generally are performed between 5 and 8 yrs of age, or later during adolescence. The decision for surgery is based on the surgeon's subjective evaluation of the lip at rest and during function, and clinicians recognize that more objective evaluation methods would be highly beneficial for the decision-making process (Brattstrom *et al.,* 2005; Trotman *et al.,* 2007a).

To address this issue, we conducted a longitudinal clinical trial (Trotman *et al*., 2007a,b,c) to examine the effects of lip revision on circumoral function as characterized by objective measures. Initial baseline findings demonstrated that patients with a repaired cleft lip had impaired circumoral movements, with lateral movements of the upper lip affected to a greater extent, and some had abnormal compensatory lower lip movements. These findings were visualized in three dimensions by methods developed by our research team (Faraway, 2004; Trotman *et al.*, 2005). Moreover, impaired circumoral movements were paralleled by abnormal force regulation of the lip muscles. Specifically, compared with control individuals, patients did not maintain normal functioning target forces as steadily, and their lower lip muscles tended to recruit forces faster, exerted excessively high peak forces, and exhibited compensatory force adjustments for decreased upper lip force.

Building on these findings, the objective of this clinical trial was to quantify the effects of lip revision surgery on circumoral movements in patients with repaired cleft lip/palate. It was hypothesized that additional (revision) surgery would increase tissue scarring and impairment. The null hypothesis (H_0) was that lip revision has no effect on facial movement. The alternative (H_A) was that lip revision further impairs facial movement.

MATERIALS & METHODS

The study design was a parallel, non-randomized clinical trial with three participant groups: (1) patients with repaired cleft lip/palate who had lip revision (revision); (2) patients with repaired cleft lip/palate who did not have lip revision (non-revision); and (3) non-cleft 'control' individuals (non-cleft). (NIH Study Section reviewers have indicated that randomization, which would have necessitated a 15-month delay for lip revision surgery, could raise potential ethical issues.) The patients in the non-revision group either declined revision surgery or were not referred for surgery, and were included for assessment of the effects of maturation without surgery. Participants were

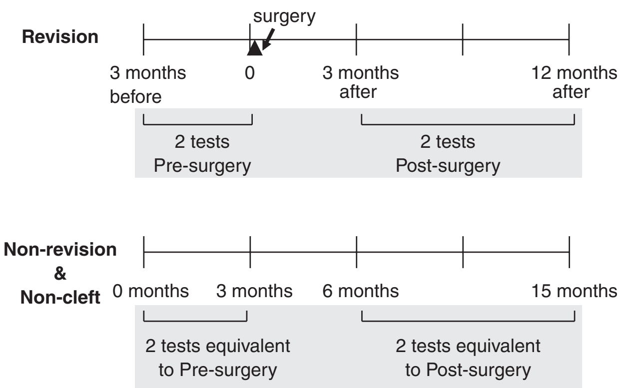

Figure 1. Test sessions. Revision patients were tested twice before and twice after surgery. Non-revision and non-cleft participants were tested at corresponding times.

screened and recruited in clinics at the University of North Carolina (UNC) School of Dentistry. The inclusion criteria were: an interest and parent willingness to participate; an ability to comprehend verbal instructions; and, specifically for the patients, a previously repaired complete unilateral or bilateral cleft lip with or without a cleft palate. The exclusion criteria were: previous orthognathic or facial soft-tissue surgery; a medical history of diabetes, collagen vascular disease, and/or systemic neurologic impairment; mental or hearing impairment such that comprehension or ability to perform tests was hampered; and, specifically for the patients, a lip revision surgery within the preceding two years.

The study purpose and protocol were explained, and informed consent and assent were obtained. Consent and HIPAA documents were approved by the UNC Biomedical Human Subjects IRB, and a Data Safety Monitoring Board was convened. The primary outcome measure for the trial was circumoral movement. Secondary outcomes were measures of lip force, EMG activity, and sensation. The results for the primary outcome measure are presented here. A sample size of 34 participants in each group was estimated, based on an anticipated difference in the change in movement between the revision and non-cleft participants. It was expected that mean facial movements would be large (50%) after lip revision, but smaller changes (10%) would be expected due to maturation in the non-cleft and non-revision groups. The sample size *per* group was sufficient to detect a large effect size (ES > = 0.80) between the revision and non-cleft groups at a 0.05 level of significance and 90% power. Blinding (masking) procedures were not possible in this study.

Participants in each group were followed longitudinally and tested at 4 times over a 15-month period (Fig. 1). Revision patients were tested at approximately 3 mos and just before surgery, and then again at approximately 3 and 12 mos after surgery. Participants in the non-revision and non-cleft groups were group-matched to the revision group and tested at similar intervals.

Intervention

Lip revision surgery was performed at the recommendation of the surgeon after consultation with the patient and parents. The surgery occurred shortly after the second test session (Fig. 1). The surgical technique has been described previously (Trotman *et al.,* 2007a).

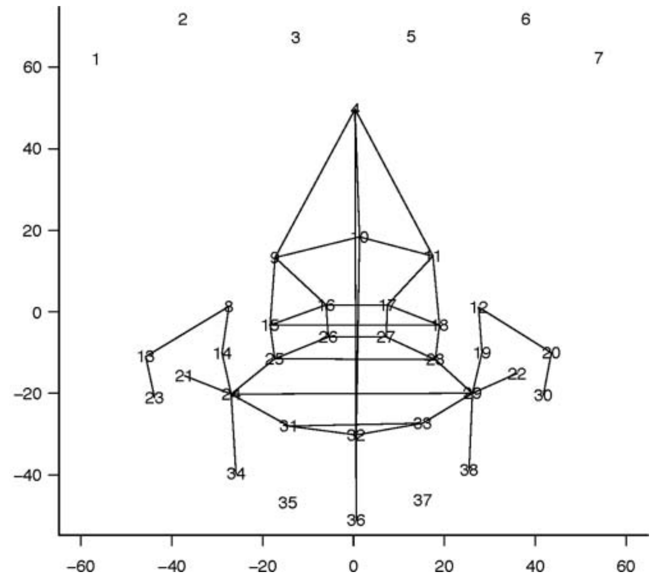

Figure 2. The 44 landmark-pairs (denoted by a line connecting 2 markers) used to develop the measurements of movement. Markers are located at the average of the rest position for the control participants.

Facial Movement Testing & Data Acquisition

We used a video-based tracking system (Motion Analysis™, Motion Analysis Corporation, Santa Rosa, CA, USA) to measure circumoral movements (Trotman *et al*., 2007a). This system tracked 38 hemispherical, 3-mm-diameter, retro-reflective markers secured to specific facial landmarks during 6 different animations—maximum smile, lip purse, cheek puff, mouth opening, and grimace, as well as natural smile. These animations represented the range of movements expected during facial expressive behavior (Darwin, 1998). Five repetitions of each animation were recorded at each test session. The raw data consisted of a timed series of 3-D vectors defined by the x, y, and z coordinate data that represented the position in space of each landmark recorded at 1/60-second intervals. The measurement of movement was the relative change in distance between 44 landmark-pairs selected to represent movements displayed by all the facial animations (Fig. 2).

Let $d_{ij}(t)$ be the distance between 2 landmarks, i and j, at time t. The relative change in the distance between these 2 landmarks from rest for a particular movement is $r_{ij}(t)=(d_{ij}(t)/(d_{ij}(0))-1$. This representation of 'relative change' had 3 advantages: (1) Effects due to facial size and shape were removed; (2) motion of the entire head had no effect on the measurement; and (3) effects due to slight variations in landmark identification between visits were minimized. The maximum relative changes in landmark-pair distances were computed. Distances that contracted were recorded as negative and those that expanded as positive, respectively.

Table. Results for Models 1 and 3

Differences between Patients with a Cleft Lip & Non-cleft Control Population					Changes due to Lip Revision (revision group only)		
Movement-type column 1	Critical Landmark-pairs column 2	Control Group column 3	Patients with a Cleft Lip column 4	Difference in Movement column 5	Before Surgery column 6	3 mos. after Surgery column 7	12 mos. after Surgery column 8
Maximum smile							
Expansion	25-28	36.2	25.8	10.4***	25.0	23.4	25.3
Contraction	16-26 & 17-27	–23.8	–13.2	–10.6***	–11.1	–12.0	–13.3*
Natural smile							
Expansion	25-28	25.8	19.5	6.3***	18.9	18.8	19.4
Contraction	16-26 & 17-27	–17.0	–10.8	–6.2***	–8.6	–9.7	–10.2
Cheek puff							
Expansion	16-26 & 17-27	25.0	14.4	10.6***	14.9	12.6	15.0 ms
Contraction	25-28	–14.4	–8.8	–5.6***	–8.5	–6.8	–7.6*
Lip purse							
Expansion	16-26 & 17-27	21.4	14.9	6.5**	15.8	13.4	14.9
Contraction	25-28	–19.2	–15.4	–3.8***	–14.6	–13.4	–14.6
Grimace							
Expansion	15-25	13.7	10.9	2.8***	10.6	11.3	11.4
Contraction	9-15 & 11-18	–13.5	–17.1	3.6**	–17.1	–14.8	–15.9 ms
Mouth open							
Expansion	25-26 & 27-28	29.8	23.3	6.5***	20.2	20.7	22.3
Contraction	24-29	–9.4	–10.0	Ns	–11.1	–11.3	–10.5

*$p \le 0.05$; **$p \le 0.001$; ***$p \le 0.0001$; ms = marginally significant at $p \le 0.07$ effect on the response for one unit change in the jaw motion.
The first column shows the predicted value of the measure for a control participant who is female, white, aged 12.5 yrs, with the median value of jaw motion and facial angle. The second shows the predicted value for a patient with the same characteristics. The jaw motion is measured by β_{jaw}, and measures the effect on the response for one unit change in the jaw motion.

Statistical Analysis

Two complementary analyses were used: (1) a critical measure analysis that used a single scalar measure to represent facial motion; and (2) a full shape analysis that retained the full shape change information in the movement.

(1) Critical Measure Analysis

For each landmark-pair, the pre-surgery data from the patients (combined revision + non-revision groups) and non-cleft individuals were subjected to a *t* test for identification of landmark-pairs that best distinguished between the two groups. Subsequent analyses were shown on these particular landmark-pairs (Table, column 2), although qualitatively similar results were obtained for other landmark-pairs of interest. To determine the effects of surgery on circumoral movements, we fit 3 linear mixed-effects models to the data for each animation.

Model 1 used the pre-surgery data and assessed the differences between the patients and non-cleft individuals:

$$y_i = \mu_0 + group_{(i)} + \beta_{jaw} jaw_i + \beta_{age} age + race_i + gender_i + \beta_{angle} angle + subject_{(i)} + visit_{(i)} + \varepsilon_i$$

Model 2 used the pre-surgery data and assessed the effect of the 'cleft-related' factors:

$$y_i = \mu_0 + group_{(i)} + \beta_{jaw} jaw_i + \beta_{age} age + cpalate_{(i)} + clip_{(i)} + maxexp_{(i)} + bonegraft_{(i)} + nasal_{(i)} + subject_{(i)} + visit_{(i)} + \varepsilon_i$$

Model 3 used data from the revision group only and assessed the effects of lip revision surgery:

$$y_i = \mu_0 + visfac_{(i)} + \beta_{jaw} jaw_i + clip_{(i)} + subject_{(i)} + visit_{(i)} + \varepsilon_i$$

y The measure of motion.

(i) Subscripted in parentheses maps case (i) to the corresponding level of the factor concerned.

group two- or three-level factor denoting group membership (grouping depends on the model).

jaw The larger of the minimum or maximum of the relative change in movement of the landmark-pair #4-36.

Age Age (yrs) at which lip revision is performed.

race A four-level factor—Caucasian, Black, Asian, Hispanic.

gender A two-level factor denoting sex.

angle The facial profile angle measurement of the participant.

subject A multi-level factor denoting participant. The number varies between models and is modeled as random effect with mean "0" and a variance to be estimated.

visit A multi-level factor denoting visit. First two visits were used in models 1 and 2. All four visits in model 3. Modeled as a random effect nested within the participant.

cpalate A two-level factor denoting the presence of a cleft palate.

clip A two-level factor denoting a bilateral or unilateral cleft lip.

maxexp A two-level factor denoting the presence/absence of maxillary palatal expansion.

bonegraft A two-level factor denoting the presence/absence of an alveolar bone graft.

nasal A two-level factor denoting the presence/absence of a nasal revision.

visfac A three-level factor. The first level denotes pre-surgery observations; the second, 3 mos after surgery; and the third, the final visit observations.

(2) Whole Shape Analysis

This shape principal components analysis incorporated dynamic time-warping and generalized Procrustes methods to produce dynamic group comparisons of the animations showing the entire face (see Appendix for description).

RESULTS

Participants were recruited from May 2001 to November 2005, and follow-up was extended to July 2007. Of the 32 non-revision patients analyzed, 21 (66%) were male, and the mean group age was 12.4 yrs (SD = 3.3). Fifty-five percent of the 34 revision patients were male, and the mean group age was 13.3 yrs (SD = 4.4). One revision patient was disqualified for non-compliance at the first test session. The mean age of the 37 non-cleft participants was 13.1 yrs (SD = 3.6), and 54% were male. The study used 'intent to treat' analyses. The predicted mean movements of the landmark-pairs for each animation represent Caucasians, females, median age (12.5 yrs), median jaw movement, and median facial angle (Table, columns 3, 4). The differences in movement between the patient and the non-cleft groups in Model 1 were significant ($p < 0.001$; Table, column 5), with substantially greater average movements for the non-cleft group.

There were no differences between the revision and non-revision groups in Model 2; however, the average movements for the maximum smile (expansion), cheek puff (expansion), lip purse (expansion), and grimace (contraction) were significantly greater in magnitude by 4% to 6% for those patients with a unilateral *vs.* a bilateral cleft lip. Model 3 gives the predicted mean longitudinal response for the revision group. No differences were found between the 2 pre-surgery means, which were averaged (Table, columns 6, 7, 8). The other variables were set to their median/baseline values. The results showed that the landmark-pairs for maximum smile (contraction) and cheek puff (contraction) demonstrated significant average changes from baseline to 12 mos after surgery, while the cheek puff (expansion) and grimace (contraction) demonstrated marginally significant ($p < 0.07$) changes. For these animations, the 12-month post-surgery scores tended to return to baseline (pre-surgery) values, but fell short of the control scores. Jaw motion had a statistically significant, but mild, effect in all three models. Over the 15-month period of follow-up, no maturational changes in movement were found in the non-revision and non-cleft groups. The models demonstrated that the among-participant variation was greatest and comparable with the group differences. The between-visit and the residual variations were somewhat smaller, but still considerable.

The results of the whole (full) dynamic shape analysis are demonstrated online (http://www.maths.bath.ac.uk/~jjf23/face/focls/) and show comparisons of mean movements between the non-cleft control individuals and patients, and, for the revision patients, comparisons of the mean movements at pre-surgery and the final post-surgery visits. Of greater interest to the clinician are the individual changes due to surgery, as demonstrated for the maximum smile landmark-pairs (Fig. 3). The horizontal and vertical dotted lines on both axes are the 'control' mean distance scores, which contracted 25%. Revision patients are superimposed. Those near the solid diagonal line—the vast majority—experienced little change due to the surgery. All patients fell in the upper right quadrant, indicating less contraction than the control individuals.

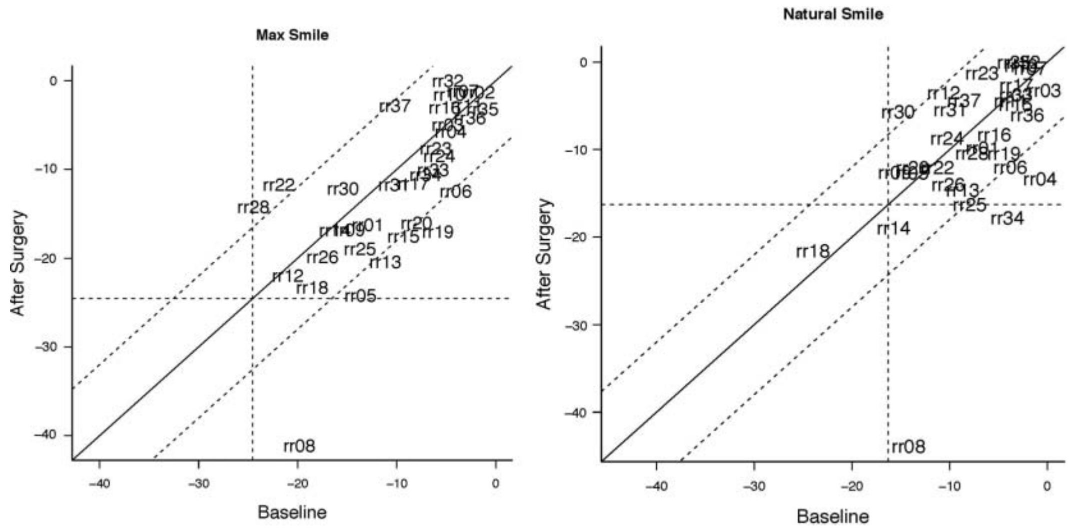

Figure 3. Changes due to surgery for the revision patients. The solid diagonal line represents no change, while the 2 parallel diagonal dotted lines represent approximately 2 standard errors' difference from no change. The horizontal and vertical lines represent the control group mean.

For the maximum smile, patient rr28 had scores equal to those of the control individuals before surgery (~ -25 along the x-axis = 25% contraction). After surgery, the scores were less than those of the control individuals (~ -15 on the y-axis = 15% contraction), suggesting increased impairment (for visualization, see http://www.maths.bath.ac.uk/~jjf23/face/focls/). Patient rr05 experienced the reverse—greater contraction after surgery. Patient rr08 had scores close to those of the control individuals before surgery, but after surgery the scores were more negative, implying more contraction than in the control individuals. Patient rr08 has a right unilateral cleft lip and demonstrated asymmetry, with greater contraction on the left side of the lip. For the natural smile (Fig. 3), the scores were closer to the control mean, with less variation than for the maximum smile.

DISCUSSION

The main finding of this trial was that, on average, lip revision did not affect facial movements, and there was some evidence of a mild improvement that led to a rejection of the alternative hypothesis. It appears that the decision for lip revision may be justified by an expectation of esthetic improvement based on the surgeon's subjective assessment of lip disability. There are, however, two important issues that should be considered. First, average change implies that some patients improved while others got worse, as was clearly demonstrated by the study findings. Thus, an objective assessment of an individual's impairment in facial soft-tissue movement would be an important measure. Moreover, the methodology presented would allow impairments to be isolated from disfigurement or problems with static lip form. Disfigurement has been shown to confound subjective evaluations of movement/function (Ritter *et al.,* 2002). The finding that movement during the maximum smile was more impaired than that of the natural smile suggests that patients' extreme movements are more affected. Presently, studies are ongoing by our research group to determine how surgeons can best use objective functional data to supplement their subjective evaluations and improve surgical outcomes.

A related factor is that the patients in this report were followed for 1 yr. Maturation during adolescence may result in an improvement or a worsening of the impairment. Evidence for maturational effects comes from studies of bony tissues following facial surgery in which changes continued well beyond 1 yr (Schubert *et al*., 1999). Longer follow-up is crucial to an understanding of the degree of adaptive changes relevant to the final outcome of lip revision, and these studies currently are being conducted. The findings presented here were those from participants attending clinics at UNC, and may not be generalizable to a broad population; however, this assessment of surgical outcomes can be used to compare the effects of different surgical techniques in clinical settings and randomized clinical trials.

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